

PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	The “Hikikomori” Syndrome: worldwide prevalence and co-occurring major psychiatric disorders. A systematic review and meta-analysis protocol
AUTHORS	Ferretti, Fabio; Pozza, A; Coluccia, Anna; Kato, Takahiro; Gaetani, Marco

VERSION 1 – REVIEW

REVIEWER	Dr. Gloria Hongyee Chan Caritas Institute of Higher Education, Hong Kong
REVIEW RETURNED	15-Sep-2018

GENERAL COMMENTS	<p>In view that a systematic review with meta-analysis which quantitatively summarizes the prevalence of the Hikikomori Syndrome (HS) is lacking, this study protocol illustrates a research to fill the research gap, which is hoped to guide future research directions and generate practice implications, so as “to inform early detection and prevention strategies in the community, and also to improve clinical practice with psychiatric patients” (on pp. 11-12 of the manuscript). It is believed that the study can bring about significance in terms of research and practice.</p> <p>A major strength of this research is the robust methodology which describes in detail the selection of studies and the analyses of the selected studies, not to mention the overall clarity of the presentation of the study protocol. Regarding the criteria for the study selection, I wish to share some opinions. It is mentioned in the study protocol that “socio-cultural influences have been believed as key factors involved in the development of this condition [HS]” (on p. 3 of the manuscript), nevertheless, to my understanding, HS appears to be less likely a “culture-bound” syndrome (on p. 3 of the manuscript) as time goes on, but appear to exist in other various places such as France, Italy, Korea, and Hong Kong. The socio-cultural influences of HS probably involve a variety of societal/cultural factors such as the societal structure (e.g., the mainstreaming culture, the labeling effects, the academic expectations imposed on students which are prominent in Asian countries), media (e.g., media enunciation when reporting the issue), school (e.g., bullying), and the family (e.g., enmeshed parent-child relationships), which have been being discussed in literature since early. Apart from selecting studies based on “the criteria proposed by Kim et al. (2012), the criteria by Teo and Gaw (2010), the criteria by Tateno and colleagues (2012) or the criteria by the Japanese Ministry of Health, Labour and Welfare published in 2010” (on pp. 6-7 of the manuscript) which are undoubtedly internationally recognized, would the eligibility criteria be broadened so as to optimize the balance between clinical and</p>
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	<p>non-clinical studies that are selected for study? Besides, the choice and adoption of the selection criteria will affect the use of keywords too (e.g., In addition to the use of keywords including “Social withdrawal”, “Hikikomori Syndrome”, “Hidden youth”, and “Severe social isolation” as stated on p. 7 of the manuscript, would there be any other possible keywords like “Socially withdrawn youth” and “Young hermits”?).</p> <p>In general, I believe that this study protocol merits publication. It would be great if a minor revision of the study protocol is undertaken before being published in BMJ Open.</p>
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REVIEWER	Jaume Vives Universitat Autònoma de Barcelona. Spain
REVIEW RETURNED	16-Apr-2019

GENERAL COMMENTS	<p>The study aims at studying the prevalence of HS, which is quite a novel syndrome worth of research.</p> <p>My major concerns are:</p> <ul style="list-style-type: none"> -I think it does not add much relevant knowledge to what has already been found in previous systematic reviews, except for the prevalence, which is quite a narrow objective which I am not sure that deserves a whole article. I would propose authors to think about a broader, more informative objective. -I don't see how knowing the prevalence of HS might "... suggest what future research should investigate" or "... inform early detection and prevention programs in the community and improve practice with psychiatric patients", as authors state in Strengths and limitations. <p>Other concerns:</p> <p>Methods</p> <ul style="list-style-type: none"> -Will you really be able to include all the languages? -You only state the keywords to be used. Boolean search for each database is not provided. -Instead of referring to several authors criteria regarding the definitions of HS, an operational definition of the authors' concept of HS should be clearly stated so that inclusion criteria of HS are clearly conveyed. -I don't think that the data planned to be extracted will allow to "... suggest what future research should investigate" or "... inform early detection and prevention programs in the community and improve practice with psychiatric patients".
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VERSION 1 – AUTHOR RESPONSE

Reviewer 1

Reviewer Name: Dr. Gloria Hongyee Chan. Institution and Country: Caritas Institute of Higher Education, Hong Kong

In view that a systematic review with meta-analysis which quantitatively summarizes the prevalence of the Hikikomori Syndrome (HS) is lacking, this study protocol illustrates a research to fill the research gap, which is hoped to guide future research directions and generate practice implications, so as “to inform early detection and prevention strategies in the community, and also to improve clinical practice with psychiatric patients” (on pp. 11-12 of the manuscript). It is believed that the study can bring about significance in terms of research and practice.

Response: We warmly thank the reviewer for her constructive comments and for appreciating our work.

A major strength of this research is the robust methodology which describes in detail the selection of studies and the analyses of the selected studies, not to mention the overall clarity of the presentation of the study protocol. Regarding the criteria for the study selection, I wish to share some opinions. It is mentioned in the study protocol that “socio-cultural influences have been believed as key factors involved in the development of this condition [HS]” (on p. 3 of the manuscript), nevertheless, to my understanding, HS appears to be less likely a “culture-bound” syndrome (on p. 3 of the manuscript) as time goes on, but appear to exist in other various places such as France, Italy, Korea, and Hong Kong. The socio-cultural influences of HS probably involve a variety of societal/cultural factors such as the societal structure (e.g., the mainstreaming culture, the labeling effects, the academic expectations imposed on students which are prominent in Asian countries), media (e.g., media enunciation when reporting the issue), school (e.g., bullying), and the family (e.g., enmeshed parent-child relationships), which have been being discussed in literature since early. Apart from selecting studies based on “the criteria proposed by Kim et al. (2012), the criteria by Teo and Gaw (2010), the criteria by Tateno and colleagues (2012) or the criteria by the Japanese Ministry of Health, Labour and Welfare published in 2010” (on pp. 6-7 of the manuscript) which are undoubtedly internationally recognized, would the eligibility criteria be broadened so as to optimize the balance between clinical and non-clinical studies that are selected for study? Besides, the choice and adoption of the selection criteria will affect the use of keywords too (e.g., In addition to the use of keywords including “Social withdrawal”, “Hikikomori Syndrome”, “Hidden youth”, and “Severe social isolation” as stated on p. 7 of the manuscript, would there be any other possible keywords like “Socially withdrawn youth” and “Young hermits”?).

Response: We warmly thank the reviewer for all these interesting points she has raised. We agree with these observations that support further our rationale related to the investigation of the world prevalence of the HS. We have added at page 4 a statement highlighting the hypothesis that the Hikikomori Syndrome may not be a culture-bound syndrome as follows “Other researchers suggested that the HS might not be a culture-bound syndrome depending on the socio-cultural context but that it may exist also outside Asian countries²²”. In addition, we have added at the same page some statements based on the suggestions provided by the reviewer to support the hypothesis that general socio-cultural factors may influence the development of the HS, as follows: “The socio-cultural features of the HS probably involve a variety of cross-cultural factors such as the social structure (e.g., the mainstreaming culture, the labeling effects, the academic expectations imposed to students which are prominent in Asian countries but also in other countries), the society’s media (e.g., media enunciation when reporting the issue), the school context (e.g., the bullying phenomenon), and the family relationships (e.g., enmeshed parent-child relationships)”.

Finally, in order to broaden the eligibility criteria and the literature search, the additional keywords she has suggested (“Socially withdrawn youth”, “Young hermits”) have been added in the search strategy.

-In general, I believe that this study protocol merits publication. It would be great if a minor revision of the study protocol is undertaken before being published in BMJ Open.

Response: We warmly thank the reviewer for appreciating our work.

Reviewer: 2

Reviewer Name: Jaume Vives. Institution and Country: Universitat Autònoma de Barcelona. Spain

The study aims at studying the prevalence of HS, which is quite a novel syndrome worth of research.

-I think it does not add much relevant knowledge to what has already been found in previous systematic reviews, except for the prevalence, which is quite a narrow objective which I am not sure that deserves a whole article. I would propose authors to think about a broader, more informative objective.

Response: We thank the reviewer for this important comment that helps use to explain better our rationale. We had to present the objectives according to the PRISMA guidelines based on the PICOS approach, the internationally recognized guidelines in conducting reviews. In addition, we think that our review adds some important knowledge to the current literature for some reasons. A first point regards the fact that the existing systematic review (Li & Wong, 2015) included only English papers searched in February 2015 and it did not provide a quantitative summary of the prevalence rates of the Hikikomori Syndrome (HS) using meta-analysis. Our review is the first quantitative systematic review conducted according to the PRISMA guidelines and it may include also the papers potentially published between 2015 and 2019. The fact that no language restriction will be applied in the search strategy may allow us to locate potential papers published in non-English languages (i.e. Japanese). The knowledge about the prevalence rates of the HS worldwide can provide an insight about the importance of the assessment of and the intervention on this phenomenon outside Japan. In addition, the existing studies conducted in the general population have produced heterogenous prevalence data, showing that the prevalence of the HS can range from approximately 0.87% to 1.2% in Japan, to 1.9% in Hong Kong to 2.3% in Korea or up to 26.66% in student populations in Japan. Thus, a quantitative summary may be important and the investigation of the reasons for this heterogeneity should also be studied (i.e. the moderator analysis). Indeed, another original point regards the fact that our review will investigate the role of some moderators including age, gender, and type of countries where the study has been conducted (Asian versus non-Asian countries).

-I don't see how knowing the prevalence of HS might "... suggest what future research should investigate" or "... inform early detection and prevention programs in the community and improve practice with psychiatric patients", as authors state in Strengths and limitations.

Response: We thank the reviewer for this comment. We agree with this. We have changed this statement in the Strengths and Limitations section and we have removed the statement "inform early detection and prevention programs in the community and improve practice with psychiatric patients". We think that the knowledge about the epidemiological aspects related to a mental condition is one of the first steps essential to understanding it. If a mental condition has a prevalence rate similar to the rates of some major disorders (e.g. anxiety disorders or personality disorders), this can suggest the need for the development of more careful detection/intervention strategies. The prevalence rates of the condition in the general population may be used as a comparator data to analyze whether a prevention strategy is effective in the future. As our review aims to assess the role of moderators such as age, gender and country, this paper can inform policymakers about which subgroups of young people may present with higher prevalence rates of this phenomenon, so they should be monitored more carefully.

Other concerns:

Methods

-Will you really be able to include all the languages?

Response: We thank the reviewer for the opportunity to provide more details about this point. No restriction regarding language will be applied. So, we will not exclude a potentially eligible paper due to the language. We believe that this point is an important aspect of the search strategy. A focus only on English papers might not allow us to locate relevant papers. We expect to find some papers published in Japanese (one of the coauthors of the review is a Japanese native speaker).

-You only state the keywords to be used. Boolean search for each database is not provided.

Response: The Boolean search for each database has been added in the Search strategy provided in the Supplementary Material. In addition, at page 7 more detailed information has been added regarding the Boolean search as follows: "The PubMed MeSH on Demand Tool allowed us to generate relevant MeSH terms. MeSH terms or keywords related to the HS concept ("Social withdrawal", "Hikikomori Syndrome", "Hidden youth", "Severe social isolation", "Socially withdrawn youth", "Young hermits") will be combined with MeSH or keywords related to prevalence ("Prevalence", "Prevalence studies", "Population-based study", "Epidemiology") through the boolean operator AND. The search procedure will be conducted during the second week of June 2019 (start date: 06/10/2019; end date: 06/16/2019) using the databases Scopus, PubMed, PsycINFO, and Web of Science. This search strategy will be used for each one of the databases".

-Instead of referring to several authors criteria regarding the definitions of HS, an operational definition of the authors' concept of HS should be clearly stated so that inclusion criteria of HS are clearly conveyed.

Response: We thank the reviewer for this suggestion. Following this comment, we have revised the eligibility criteria related to the HS definition and clearly added an operational definition of the concept as follows: "Studies will be included if [...] they investigated the HS and conceptualized it as an independent psycho-sociological condition including the following features: (1) Spending most of the day and almost every day at home, (2) Marked and persistent avoidance of social situations, (3) The social withdrawal and avoidance interferes significantly with the person's normal routine, occupational (or academic) functioning, or social activities or relationships, (4) The person perceives the withdrawal as ego-syntonic, (5) In individuals under age 18 years, the duration is at least 6 months, (6) The social withdrawal is not better accounted for by another mental disorder (e.g, Social Anxiety Disorder, Major Depressive Disorder, Schizophrenia, or Avoidant Personality Disorder). Despite numerous criteria were proposed in the literature to define the HS including those suggested by Kim et al.,⁴² by Tateno et al.⁴³ or the criteria by the Japanese Ministry of Health, Labour and Welfare published in 2010,⁴⁴ we have used the above-mentioned criteria following the definition provided by Teo and Gaw^{13,32} who identified the most recurrent clinical features across papers published in peer-review journals."

-I don't think that the data planned to be extracted will allow to "... suggest what future research should investigate" or "... inform early detection and prevention programs in the community and improve practice with psychiatric patients".

Response: Following the reviewer's suggestion, we have removed this statement as previously states.

VERSION 2 – REVIEW

REVIEWER	Jaume Vives Universitat Autònoma de Barcelona (Spain)
REVIEW RETURNED	08-May-2019

GENERAL COMMENTS	<p>I appreciate that authors have addressed the reporting of the operational definition of the syndrome.</p> <p>Issues not addressed:</p> <p>-As I mentioned in the previous review, I think it does not add much relevant knowledge to what has already been found in previous systematic reviews, except for the prevalence, which is quite a narrow objective which I am not sure that deserves a whole article. I would propose authors to think about a broader, more informative objective. I did not see anything new in this regard in the reviewed manuscript.</p> <p>-As I mentioned in the previous review, I still don't see how prevalence "... may suggest what future research should investigate" or "Identifying prevalence rates may inform early</p>
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	<p>detection and prevention programs in the community” as authors state in the Strengths and limitations. Again, I did not see anything new in this regard in the reviewed manuscript.</p> <p>-Search strategy: Boolean operators are not reported in detail. Only an AND is reported. Further, what fields will be used to search? Nevertheless, regarding the search strategy, I am not sure that including AND followed by Prevalence”, “Prevalence studies”, “Population-based study”, “Epidemiology” is a good idea. This search may exclude studies reporting prevalence while not specifically stating it.</p> <p>-I also found a kind of Inconsistency. In the Abstract it is stated that “No restriction about design or language will be applied”, while in the Conclusion section “The potential strengths of the study will be that the existing previous PRISMA systematic review focused mainly on English papers, while most research was conducted in Japan and some papers were published in Japanese”.</p>
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VERSION 2 – AUTHOR RESPONSE

Reviewer: 2

Reviewer Name: Jaume Vives

Institution and Country: Universitat Autònoma de Barcelona (Spain)

I appreciate that authors have addressed the reporting of the operational definition of the syndrome.

Issues not addressed:

-As I mentioned in the previous review, I think it does not add much relevant knowledge to what has already been found in previous systematic reviews, except for the prevalence, which is quite a narrow objective which I am not sure that deserves a whole article. I would propose authors to think about a broader, more informative objective. I did not see anything new in this regard in the reviewed manuscript. Response. Following the Reviewer’s comment, we have changed the overall focus of the review by adding a further broad objective, i.e. the co-occurrence of HS and each psychiatric disorder defined by any version of DSM or ICD. We have added the investigation about which psychiatric disorders (according to any version of these two classification systems) are the most commonly co-occurring in young people with HS. Therefore, a new objective has been added in the text, as follows: “[...] to assess the co-occurrence between HS and each psychiatric disorder defined by the criteria of any version of the DSM or ICD in any clinical samples with psychiatric disorders. To provide a broad overview of the co-occurrence between HS and psychopathology, we will consider any psychiatric disorder based on any version of these two classification systems which are the most internationally used systems. Following the clinical observations and hypotheses proposed by previous authors,^{13,21,24-25,32} we expect that the highest co-occurrence rates can be found for psychosis, unipolar depressive disorders, social anxiety disorder, schizoid personality disorder, avoidant personality disorder, post-traumatic stress disorder, and Internet/game addiction.”. The paper title, the eligibility criteria, the search strategy, the effect size calculation and some statements in the Discussion section have been revised to reflect this additional broader focus accordingly.

-As I mentioned in the previous review, I still don’t see how prevalence “... may suggest what future research should investigate” or “Identifying prevalence rates may inform early detection and prevention programs in the community” as authors state in the Strengths and limitations. Again, I did not see anything new in this regard in the reviewed manuscript. Response. Following the Reviewer’s

suggestion, we have removed the above-mentioned bullet points in the Strengths and Limitations section. We have replaced them with other bullet points related to the new broader scope of the review. As requested by the Reviewer, we have eliminated from the Rationale for the present study paragraph the statement related to the fact that identifying prevalence rates may inform early detection and prevention programs in the community. We have highlighted that the investigation of the psychiatric disorders co-occurring in the Hikikomori Syndrome is a new aspect, unexplored by previous reviews. To support the originality of this review we have added the following sentence in the Rationale for the present study paragraph: "A systematic review with meta-analysis summarizing the prevalence rates of this condition and the co-occurrence of psychiatric disorders has not been conducted. None of the previous reviews summarized the co-occurring rates of psychiatric disorders in HS. In order to have a more comprehensive understanding of the clinical picture of this condition, it seems important to investigate which psychiatric disorders are most frequently associated with it.". We think that a comprehensive review of the co-occurrence rates between Hikikomori Syndrome and each psychiatric disorder defined by the DSM or ICD may add some knowledge to the current understanding of this condition because a quantitative summary about this aspect is lacking in the literature.

-Search strategy: Boolean operators are not reported in detail. Only an AND is reported. Further, what fields will be used to search? Nevertheless, regarding the search strategy, I am not sure that including AND followed by Prevalence", "Prevalence studies", "Population-based study", "Epidemiology" is a good idea. This search may exclude studies reporting prevalence while not specifically stating it. Response. Following the Reviewer's comment, we have changed the search strategy by adding different Boolean operators and new search terms which better reflect the broad scope of the review. Please, see the Supplementaru material where a detailed overview is provided. In addition, we have removed the search terms "Prevalence", "Prevalence studies", "Population-based study", "Epidemiology", as requested by the Reviewer.

-I also found a kind of Inconsistency. In the Abstract it is stated that "No restriction about design or language will be applied", while in the Conclusion section "The potential strengths of the study will be that the existing previous PRISMA systematic review focused mainly on English papers, while most research was conducted in Japan and some papers were published in Japanese". Response. Following the Reviewer's suggestion, we have removed the above-mentioned statement in the Conclusions section to avoid inconsistency.